

# Tongue tie

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## Common problem or old wives' tale?

The resurgence of interest in breast feeding has been accompanied by a lively debate about the significance of "tongue tie" or ankyloglossia. Symptoms attributed to tongue tie include nipple pain and trauma, difficulty in the baby attaching to the breast, frequent feeding, and uncoordinated sucking. These problems may result in the mother deciding to terminate breast feeding prematurely, slow weight gain for the baby, and even hypernatraemic dehydration. Speech defects have also been attributed to tongue tie. Strong views have been expressed by many eminent authors on the subject (box 1).

This paper reviews what is known about tongue movements and the significance and treatment of tongue tie. It is based on two literature reviews, one conducted on behalf of NICE<sup>1,2</sup> by one of us (MR) and updated by further searches of published and grey literature and conference abstracts. The publications reviewed for this paper are summarised in table 1.

As our review found little high quality objective evidence, we begin by making explicit the personal experience and bias with which we commenced the review. One of the authors (MR) felt that

tongue tie is an important issue—she experienced pain for many weeks while breast feeding her first child, who exhibited features said to be typical of tongue tie, and has since discussed this issue widely with lactation specialists and women having similar problems. The other (DH) accepted that ankyloglossia occurs in dysmorphic infants<sup>10</sup> and occasionally in otherwise normal babies,<sup>11</sup> but was sceptical about the high prevalence of the condition now being described by several authors.

### ANATOMY AND PHYSIOLOGY

The tongue is a highly mobile organ made up of longitudinal, horizontal, vertical, and transverse intrinsic muscle bundles. The extrinsic muscles are the fan-like genioglossus which is inserted into the medial part of the tongue and the styloglossus and hyoglossus into the lateral portions. The sub-lingual frenulum is a fold of mucosa connecting the midline of the inferior surface of the tongue to the floor of the mouth. Tongue tie is the name given to the condition arising when the frenulum is unusually thick, tight, or short. There are many variations and differing degrees of severity (fig 1).

The movements of the tongue during infant feeding have been studied by cine-radiography and more recently by ultrasound.<sup>12,13</sup> Ultrasound reveals some similarities between the movements made by the baby when either breast or bottle feeding,<sup>14</sup> but also some important differences.<sup>15</sup> The tongue is projected further forward in breast feeding<sup>16</sup> and the human nipple elongates during each suck in a way that an artificial teat cannot do.<sup>14</sup> During feeding, the artificial teat, or the nipple together with some breast tissue, is held fully in the mouth with the tongue covering the lower gum ridge. The nipple is protected from damage and pain at the back of the baby's mouth.<sup>16</sup> The baby's lower jaw is then elevated, compressing the artificial teat, or the breast immediately behind the nipple, while the front of the tongue moves up to aid the expression of milk. In breast feeding, this is by compression of the milk ducts under the areola. A wave of upward movement of the medial part of the tongue progresses backwards, and the expression of the milk is further facilitated by negative pressure generated by downward movement of the back of the tongue and the lower jaw and, in breast feeding, by the active expulsion of milk once the let down occurs.

In coordinated feeding, the sucking, swallowing and breathing movements follow in a 1:1:1 sequence. This can take several days to become established in healthy full term infants. In pre-term infants and in some term infants a variety of poorly coordinated feeding movement patterns are observed and sometimes persist.<sup>17</sup> Antenatal

### Box 1: Quotes from the past

"In observing a very large series of newborn babies, we have never seen a tongue that had to be clipped" (McEnery and Gaines, Chicago, 1940)

"While tongue tie is not nearly as common as members of the public believe, nevertheless a genuine case is occasionally seen and the condition is not entirely mythical although surrounded by an aura of superstition and old wives' tales" (Cullum, UK, 1959)

"Tongue tie...has been described as a myth of hoary antiquity...but it is probably wrong to suggest that it never causes symptoms. A case is reported in which a tight fraenum ruptured spontaneously during feeding...this baby remained a slow feeder and...[had not been] disabled by his tongue tie" (Smithells, London, 1959)

"Tongue tie is a rare but definite congenital deformity" (Browne, London, 1959)

"Tongue tie is a rare cause of dysarthria, though it is often blamed for slow speech development...most patients who have real limitation of movement as a result of tongue tie have a history of difficult milk feeding" (Ingram, Edinburgh, 1968)

"I have never seen feeding difficulties in the first year resulting from tongue tie and I doubt whether it is ever necessary to carry out an operation on it till the age of two or three...There are still doctors who cut the frenulum in the newborn period. This is always wrong" (Illingworth, Sheffield, 1982)

"Tongue tie where the tongue is forked can, very rarely, add to the baby's difficulties in taking the breast with poor protractivity" (Gunther, UK, 1970)

"To some extent tongue tie is normal in every newborn baby and it should rarely interfere with either sucking or later speech development" (Davies *et al*, UK, 1972)

"True tongue tie is a very rare condition. This condition has been over-diagnosed in the past because of the failure to recognise that the frenum passing from the tongue to the floor of the mouth is normally short in the newborn...Only in infants with severe limitation of the tongue movement and inability to suck is division of the frenum indicated" (Turner, Douglas, and Cockburn, UK, 1988)

**Table 1** Tongue tie; review of literature

Author(s)	Number and age group studied	How cases were identified	Type of study and intervention	Results
Messner <i>et al</i> <sup>3</sup>	Examined 1041 newborns. Identified 50 TT cases (4.8%). M:F ratio 2.6:1. 36 cases of TT enrolled and 36 controls without TT	Screened by one doctor, confirmed by one colleague	Observational follow up study but no intervention	30/36 TT cases and 33/36 controls breast fed to 2 months ( $p=0.29$ ). 9 cases and 1 control experienced breast feeding difficulties
Hogan <i>et al</i> <sup>4</sup>	Examined 1866 babies. Identified 201 TT cases (10.7%). M:F ratio 1.6:1. 44% TT cases had problems feeding. 57 TT babies entered study (40 breast fed and 17 bottle fed)	Photos to assist staff in postnatal checks	Randomised to immediate frenulotomy or support by lactation counsellor, at mean age 20 days (3–70), median age 14 days	TT cases treated by frenulotomy; 27/28 marked improvement. Counselling cases managed conservatively; 1/29 improved
Ballard <i>et al</i> <sup>5</sup>	Examined 2763 breast fed in-patient babies and 273 attenders at lactation clinic. M:F ratio 1.5:1. Identified TT in 3.2% in-patients and 12.8% clinic attenders	One observer examining all babies. ATLFF used	123 cases underwent frenulotomy at age 1–2 days	Latch improved in all, pain scores fell significantly
Ricke <i>et al</i> <sup>6</sup>	Examined 3490 babies, identified 148 TT cases (4.24%). M:F ratio 2.3:1. Enrolled 49 TT babies for study with 2 matched non-TT breast fed babies as controls	Nurses assisted by photos, ATLFF by team	Observational study, no intervention	Mothers of TT babies three times more likely to give up breast feeding by one week; however, 80% TT breast feeding well at one week. TT and non-TT breast feeding in equal numbers at 1 month. Mothers with TT babies reporting more pain at one month but not statistically significant. Small numbers so type II errors possible
Ramsay <sup>7</sup>	Case series	Referrals to paediatric surgeon	Measured nipple tip to hard soft palate junction by ultrasound, pre- and post-frenulotomy	Distance changed from 7.99 mm to 6.49 mm. Milk transfer increased from 3.3 to 7.2 ml/min. At least 7 day interval between frenulotomy and 2nd measurement
Messner and Lalakea <sup>8</sup>	Case series of speech problems: 30 children age 1–12	Measured tongue protrusion and inter-incisal distance	Frenulotomy	Speech improved. 25 mothers had tried to breast feed; 21 said no problems
Fernando <sup>9</sup>	Case series, $n \geq 200$	Various; majority presenting with speech disorders	Frenulotomy	Improved to varying degree. 20% had history of BF problems: 80% did not

TT, tongue tie; ATLFF, Assessment Tool for Lingual Frenulum Function.

ultrasound studies show that mouth and tongue movements are already well developed in association with intra-uterine yawning and crying.<sup>18–19</sup>

## HYPOTHESES

Review of the literature and expert opinion gave rise to the following hypotheses:

- Tongue tie is a definable condition
- Tongue tie affects 3–4% of infants
- The tight frenulum prevents the infant from getting the tongue over the lower lip and gum ridge and therefore can cause feeding problems, particularly affecting breast feeding, leading to pain for the mother and poor infant weight gain; it can also affect bottle feeding
- The impact of a tight frenulum varies between mother–baby dyads
- A tight frenulum can also cause problems in older children and adults, involving speech, dental hygiene, licking ice cream, and French kissing

- Division of the frenulum (frenulotomy) is a low risk effective treatment
- The condition is genetic

## CASE DEFINITION

Can tongue tie be defined—and to what extent do individual observers agree on the diagnosis? The length of attachment of the frenulum varies widely. In some babies it extends to the tip of the tongue. There may be an indentation of the anterior edge, referred to as a heart shaped tongue. The appearance of the tongue is not sufficient on its own to make a diagnosis, as the thickness and elasticity of the frenulum also vary widely and affect the extent to which normal tongue movements are inhibited (see fig 1).

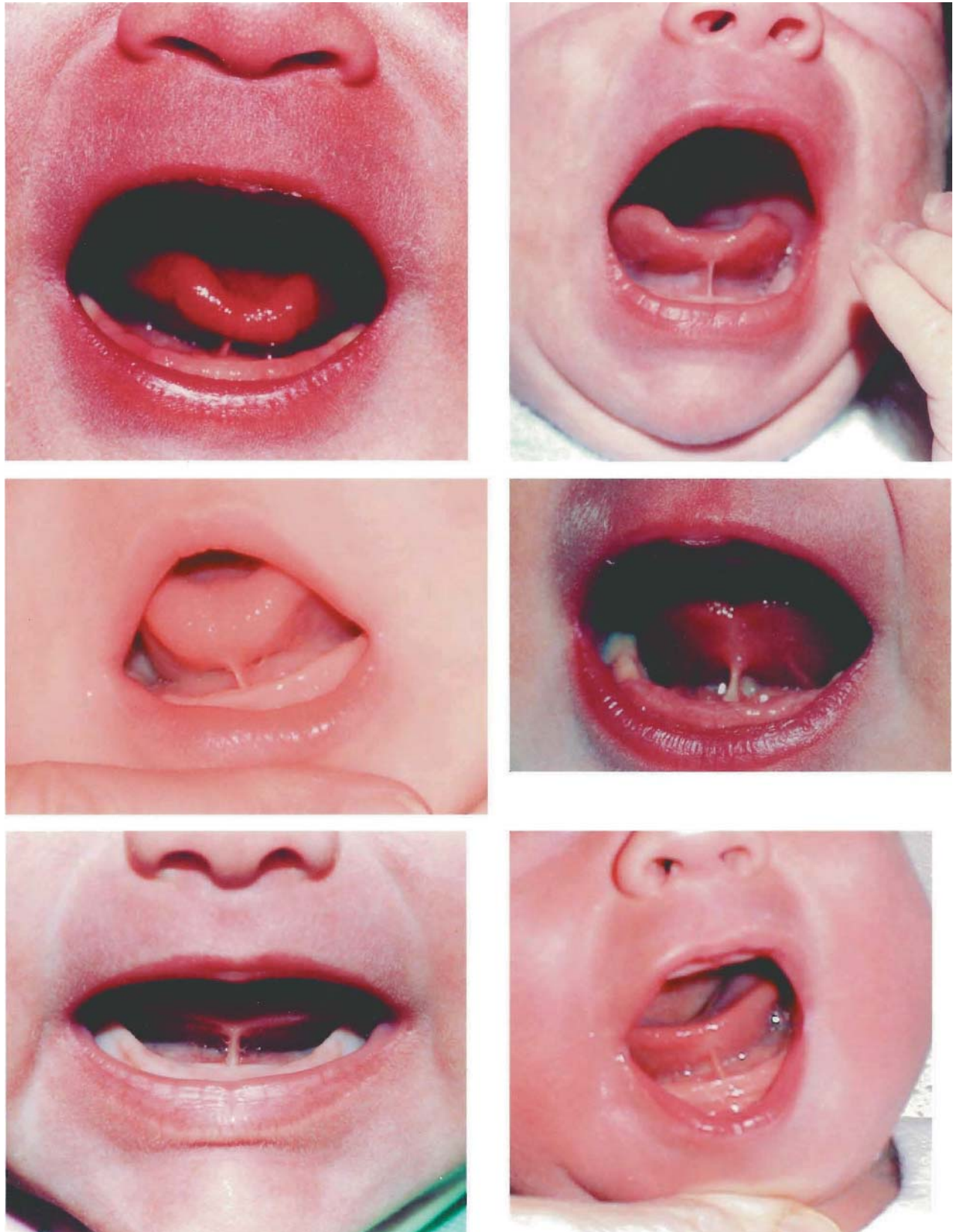
In four published studies of tongue tie in babies, the initial selection of possible cases from the whole newborn population was based only on appearance and was done by one individual (Ballard and colleagues<sup>5</sup>), or by one individual with confirmation of positive diagnoses by one other observer (Messner and colleagues<sup>3</sup>), or accomplished by providing staff with a

selection of photos (Hogan and colleagues,<sup>4</sup> Ricke and colleagues<sup>6</sup>). We have not found any formal data on observer agreement or variation in this process. The method of selection in the study by Masaiti and Kaempf<sup>20</sup> is unclear but cases were probably selected on the basis of breast feeding problems. Two authors (Messner,<sup>3</sup> Ricke<sup>6</sup>) aimed to reduce bias by trying to avoid specific mention of tongue tie to mothers but acknowledge that this was difficult.

All authors agreed that function is more important than appearance and Hazelbaker designed an Assessment Tool for Lingual Frenulum Function (ATLFF).<sup>21</sup> Ballard used this tool but did not examine inter-rater reliability. Ricke *et al* found that the inter-rater agreement using the ATLFF was only moderate and that many infants did not fit in any of the categories defined by Hazelbaker.

## BIRTH PREVALENCE OF TONGUE TIE

There is agreement among authors that tongue tie is found in around 3–4% of babies, with the exception of Hogan *et al*



**Figure 1** Six examples of babies diagnosed as having tongue tie, showing the variation in the thickness and insertion of the frenulum (reproduced with kind permission from Carolyn Westcott, Princess Anne Hospital, Southampton).



who report 10.7%. All report a male preponderance with ratios varying from 1.5:1 to 2.6:1.

### THE IMPACT OF TONGUE TIE

Maternal pain during feeding, sometimes accompanied by trauma, and difficulty in the baby taking the breast, are the main breast feeding problems attributed to tongue tie.<sup>3-6</sup> Attributing pain during breast feeding to tongue tie is not straightforward, however, since pain is a common problem that can result from several other causes, including attachment problems unconnected with tongue tie, and infection.<sup>16 22-24</sup> Ricke and colleagues<sup>6</sup> reported that more tongue tied infants than controls were bottle fed at one week but there was no significant difference at one month, though attrition meant that numbers were small. Messner *et al*<sup>3</sup> found no difference in the rate of breast feeding between tongue tied infants and controls at 2 months but a significant difference in the numbers of mothers reporting problems with breast feeding.

Ramsay<sup>7</sup> measured the distance from nipple tip to the junction of the hard and soft palate by sub-mental ultrasound. The distance decreased from 7.99 mm ( $\pm 2.80$ ) to 6.49 mm ( $\pm 1.87$ ) seven days after frenulotomy. This change, though statistically significant, is small and its practical significance is unknown. The tongue movements were said to become "more normal".

### VARIATION BETWEEN DYADS

Hogan and colleagues<sup>4</sup> found that more than half of babies with tongue tie had no problems breast feeding but could not show any correlation between severity of tongue tie and feeding difficulty. This is perhaps surprising, but it may be that only a small shift in positioning on the breast is sufficient to eliminate pain and improve feeding. Ricke and colleagues<sup>6</sup> reported that 80% of tongue tied infants were breast feeding successfully at one week. It is of interest that in two case series of older children presenting with speech difficulties and other problems attributed by the authors to tongue tie, 21/25 mothers (Messner and Lalakea<sup>8</sup>) and 80% of an unspecified number (Fernando<sup>9</sup>) who were asked about breast feeding reported no significant difficulties.

### OTHER PROBLEMS

Several case series report a range of other problems in older children associated with ankyloglossia—speech defects, difficulty in licking the lips or in kissing, dribbling, etc. These are difficult to evaluate as the authors do not give details of the catchment population, referral patterns, or detailed criteria for inclusion in the series.

### INTERVENTION

All authors agree that frenulotomy in the newborn is a low risk minor procedure, performed without anaesthetic. The presence of the deep lingual vein just lateral to the midline means that significant venous bleeding could occur if technique is not meticulous but we found no reports of serious adverse events. In older children the procedure needs an anaesthetic and sometimes a frenuloplasty, which carries some risk of scarring.

Ballard and colleagues<sup>5</sup> reported a marked fall in maternal pain scores after the procedure. Hogan and colleagues<sup>4</sup> randomised their cases to immediate or delayed intervention and found that frenulotomy was much more effective than advice from a lactation counsellor. They reported dramatic and rapid, often immediate, improvement after the procedure in most of their cases; improvement was noted in 95% of babies. The measurement of outcomes was not blinded. The precise criteria for improvement were not specified. Improvement was not always immediate, but this could be due to the need for sore nipples to heal or for the baby to re-learn optimal patterns of suckling.

In case series of older children and adults, some striking improvements were noted after surgery, but many of the children showed only gradual or modest improvement, particularly where the articulation of speech was concerned; these children often needed continuing speech therapy and this was attributed to the need to un-learn established patterns of articulation.<sup>9</sup> The absence of any comparison or control cases makes these reports impossible to evaluate and we found no comparative studies or randomised trials addressing the role of tongue tie or frenulotomy in older children.

### IS ANKYLOGLOSSIA INHERITED?

To define the inheritance of a condition, a robust case definition is needed, but tongue tie varies markedly in severity and is not an all-or-none condition. When an anomaly is identified in a newborn infant, the family searches its collective memory for other similar cases but, in the case of tongue tie, it would be impossible to assess the validity of that diagnosis in retrospect. None of the studies we reviewed considered these issues and none had gathered systematic family data across a number of families with and without the condition. Notwithstanding the comments made in several papers, no conclusions can currently be drawn about family history.

### AN OVERVIEW

Tongue tie is at first glance a minor issue, but from the results of the only randomised controlled trial yet conducted, Hogan and colleagues<sup>4</sup> suggest that at least 3% of newborns (57/1866) would benefit from frenulotomy and that this would increase the rate of continuing breast feeding. Most of the literature on tongue tie has been in connection with breast feeding; however, Hogan *et al* report that of the 57 babies in their study who benefited from frenulotomy, 40 were breast fed but 17 were artificially fed. If they are correct, this is a very common congenital anomaly that affects both breast fed and bottle fed babies, and up to 18 000 such procedures should be performed each year in the UK. It is therefore important to ask whether the evidence supports that rate of intervention and to scrutinise the evidence with a particularly critical eye.

There were a number of methodological problems with most of the studies we reviewed. These included:

- Inadequate assessment of inter-observer reliability of the initial diagnosis, the dynamic assessment of feeding and the maternal symptoms
- Ethical and practical difficulties in concealing the suspected diagnosis from the mother, thus potentially introducing a bias by raising the expectation of breast feeding problems and of improvement from intervention—this is, however, a common limitation in most studies of breast feeding problems
- Poorly defined outcome measures; it is particularly difficult to establish an objective assessment of improvement, when the primary outcome measure is reduction in maternal pain during breast feeding
- The dilemma of when to assess and intervene for tongue tie; if done very early, before breast feeding is established, as in the Ballard *et al* study, improvements may be wrongly attributed to the procedure (because suckling efficiency improves over the first few days and weeks<sup>13</sup>), but if done later (as in Hogan *et al*), many mothers may already have sore nipples or have given up breast feeding.

### CONCLUSIONS

We began this review by stating our personal bias. While DH confesses to still being somewhat more sceptical than MR, we are in complete agreement on the following conclusions:

- Individual case histories suggest that some babies do have a tight frenulum (tongue tie) which can inhibit breast

feeding and, in some cases, bottle feeding as well

- Although frenulotomy is a simple low risk procedure, it should be carried out only by those who have been trained in the procedure<sup>25</sup>
- It can be justified only if it is likely to lead to significant improvement in the comfort and the continuation of breast feeding, or of other longer term problems for the child
- We do not know the true prevalence of significant tongue tie
- There is no evidence one way or the other about inheritance
- On current evidence, there is no justification for actively searching for tongue tie during routine examination, but when mothers are having difficulty in breast feeding this should be considered as one of several possible causes
- The diagnosis should rest primarily on observation and analysis of feeding difficulties rather than the static appearance of the tongue
- It may be wise to be particularly cautious in making this diagnosis in the first two or three days before lactation is established
- The problem is of sufficient interest and importance to merit further studies of both breast and bottle fed babies, in which more precise case definition, measures of inter-observer reliability of pre- and post-intervention assessment, and ultrasound imaging are likely to play a key role
- Given the evidence that breast feeding has many advantages for both mother and baby, funding should be sought for carefully planned definitive studies on the issue.

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Parental consent was obtained for publication of the babies in figure 1

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#### Endocrinology

## Children with diabetes benefit from exercise

J I Wolfsdorf

Commentary on the paper by Massin *et al* (see page 1223)

How much physical activity do children require to obtain beneficial health and behavioural effects? The recent report concerning the effects of regular physical activity on health and behavioural outcomes in 6–18 year old youth recommends that

school age youth should participate daily in at least 60 minutes of moderate to vigorous physical activity that is developmentally appropriate, enjoyable, and involves a variety of activities.<sup>1</sup> There is strong evidence for beneficial effects of physical activity on: musculoskeletal and

cardiovascular health, adiposity in overweight youth, and blood pressure in mildly hypertensive adolescents. Physical activity also has a beneficial effect on anxiety, depression, and self-concept. The 60 minutes or more of physical activity can be achieved in a cumulative manner in school during physical education, recess, intramural sports, and before and after school programmes.

Exercise requires considerable alterations in fuel metabolism and presents unique challenges for the person with type 1 diabetes mellitus (T1D).<sup>2</sup> During the first 5–10 minutes of moderate intensity exercise, skeletal muscle glycogen is the major fuel for working muscle. With increasing duration of exercise, plasma glucose and non-esterified fatty acids (NEFA) predominate, and to meet the increased demand for